PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	The trajectory to diagnosis with pulmonary arterial hypertension: a qualitative study
AUTHORS	Ian Armstrong, Nikki Rochnia, Carl Harries, Sarah Bundock and Janelle Yorke

VERSION 1 - REVIEW

REVIEWER	Wendy Gin-Sing Pulmonary Hypertension Clinical Nurse Specialist Hammermsith Hospital London W12 0HS
REVIEW RETURNED	24/01/2012

GENERAL COMMENTS	This is a very well written and interesting article addressing the patient experience in this rare disease and emphasises the need for considering pulmonary hypertension as a differential diagnosis for breathless patients

REVIEWER	Dr Sara Booth Macmillan Consultant in Palliative Medicine Cambridge University Hospitals NHS Foundation Trust I have no competing interests.
REVIEW RETURNED	24/01/2012

GENERAL COMMENTS	This study is important, was carefully and thougtfully carried out and has been beautifully written up so that it will be easily read. It is deeply moving to read of patients' experiences and this should increase the impact of the findings. It makes an important recommendation about the investigation of undiagnosed dyspnoea, and it is to be hoped that the NHS will soon begin to get the message that cancer is not the only disease where rapid diagnosis can prevent/reduce morbidity and mortality and where empathetic as well as expert care is central to good management. THe pictures of the illness trajectory are interesting and important to have (to visualise what is mentioned in the text) but quite hard to understand. Congratulations to the authors. Note there are some typos e.g. in the abstract where key words are not separated, in box 3 where anxiolytic is incorrectly spelt.

REVIEWER	Dr Marilyn Kendall, Senior Research Fellow, University of Edinburgh, UK No competing interests
REVIEW RETURNED	09/02/2012

THE STUDY	more detail needed on methods
REPORTING & ETHICS	the study has ethical approval from the charitable organisation and the university ethics committee, but does not mention NHS ethical committee approval. I do not know whether or not this is needed for studies that recruit via charitable organisations.
GENERAL COMMENTS	Thank you for the opportunity to review this paper. Overall is it well written and clearly structured. However I do have a number of suggestions and comments, mostly concerning the methods, which are my area of expertise. I hope you will find them helpful. 1) It seems that the data reported here may be only part of the whole study data (on the experience if living with PAH ?). If this is so, it needs to be more clearly explained and justified why this part of the data from 30 interviews warrants a separate paper. 2) Sample. The 30 participants were all recruited from the Pulmonary Hypertension Association. The implications of this should be discussed in the strengths and limitations section. More detail on how participants were selected and approached in order to gain a "maximum variation sample" (p. 17) would be helpful in interpreting the results. What was the justification for completing 30 interviews rather than any other number? It might be helpful to have Table 1 as an appendix, and to include details of ethnicity, and a general overview of the sample in the methods section. 3) Data generation. Why were interviews undertaken rather than eg focus groups? Why were only patients interviewed rather than also including family carers and GPs or other health professionals? I would have liked more detail of and explanation for the use of pictorial representations (or a reference for their use), and more detail on how the individual interviews were conducted. Were negatice cases sought? 4) Data analysis. More detail is needed on how, when and by whom the data was analysed and synthesised. Were any software packages used? How did these themes develop (rather than others that you mention such as "frustration, anger and uncertainty" p.8)? 5) Results and discussion. The results section is well laid out with a good use of illustrative quotations from a range of participants. The discussion section could then consider these accounts in comparison to those known from the literature for people with a range of other conditions, in order to highlight what is similar/different in a journey to diagnosis with PAH, and what the implicatipons of this may be, and what research is needed. Andersons' model could be used in this section. 6) Strengths and limitations. Given the claims made regarding the role of medical professionals in the patients' journey to a diagnosis of PAH, the exclusion of their views from the study has implications for this and future research. See also above on sampling strategy.

	The reservations expressed regarding the nature of qualitative data perhaps convey a misapprehension of what qualitative research can do. Working, as it can only do, with the pre-interpreted domain of understanding, its strength lies in its ability to explain people's perceptions and understandings of their experience, and the meanings they attach to it, which are what health professionals have to work with, regardless of "what really happened". Consequently I feel the method was appropriate to the research question, not a limitation on it, and contributed to its strength in starting to produce research knowledge of this area of patient experience. 7) Conclusions. In the abstract the first line should be replaced with a sentence answering the research question ie describing the patients experience of the trajectory, and pulling out the main implications of it for future research and consideration (since it can be problematic to make the leap from one qualitative study to recommendations for changes in policy and practice).

VERSION 1 – AUTHOR RESPONSE

We would like to thank the reviewers for their time and comments. We found the comments to be both encouraging and constructive.

Reviewer: Wendy Gin-Sing
Pulmonary Hypertension Clinical Nurse Specialist
This is a very well written and interesting article addressing the patient experience in this rare disease and emphasises the need for considering pulmonary hypertension as a differential diagnosis for breathless patients.

Thank you for your review and comments.

Reviewer: Dr Sara Booth
Macmillan Consultant in Palliative Medicine
Cambridge University Hospitals NHS Foundation Trust
I have no competing interests.

This study is important, was carefully and thoughtfully carried out and has been beautifully written up so that it will be easily read. It is deeply moving to read of patients' experiences and this should increase the impact of the findings. It makes an important recommendation about the investigation of undiagnosed dyspnoea, and it is to be hoped that the NHS will soon begin to get the message that cancer is not the only disease where rapid diagnosis can prevent/reduce morbidity and mortality and where empathetic as well as expert care is central to good management.

Thank you for this comment – we also hope that this paper begins to shed light on the importance of thoroughly investigating unexplained dyspnoea when cancer is ruled out.

THe pictures of the illness trajectory are interesting and important to have (to visualise what is mentioned in the text) but quite hard to understand.
We have added text to accompany the pictures
Congratulations to the authors.

Thank you

Note there are some typos e.g. in the abstract where key words are not separated, in box 3 where

anxiolytic is incorrectly spelt.

Typos now corrected

Reviewer: Dr Marilyn Kendall, Senior Research Fellow, University of Edinburgh, UK
No competing interests

The study has ethical approval from the charitable organisation and the university ethics committee, but does not mention NHS ethical committee approval. I do not know whether or not this is needed for studies that recruit via charitable organisations.

Thank you for this point. NHS ethical approval was not requested since no participants were recruited through NHS means. All research contacts were made through the PHA UK membership. No interviews or other research activities were conducted on NHS premises or using any NHS resources. PHA UK has conducted survey research where questionnaires have been posted to members and that type work also required no NHS ethical approval. However, PHA UK recognises that applying for NHS REC approval for future projects may represent good practice as a method of providing external critique and assurances.

Thank you for the opportunity to review this paper. Overall is it well written and clearly structured. However I do have a number of suggestions and comments, mostly concerning the methods, which are my area of expertise. I hope you will find them helpful.
1) It seems that the data reported here may be only part of the whole study data (on the experience if living with PAH ?). If this is so, it needs to be more clearly explained and justified why this part of the data from 30 interviews warrants a separate paper.

This is correct and we agree that this could be made clearer in the methods section rather than the reference made in the results on page 8. Our intension was to prepare one manuscript relating to the experience of living with PH but the participants described such vivid stories of their pre-diagnostic phase it was agreed that this warranted a separate paper. New knowledge has surfaced from this – notably the need for clinical guidelines for the investigation of unexplained dyspnoea, across all age groups, when more common diagnoses are ruled out. This approach is supported by the comments from the clinical expert reviewers.

2) Sample. The 30 participants were all recruited from the Pulmonary Hypertension Association. The implications of this should be discussed in the strengths and limitations section. More detail on how participants were selected and approached in order to gain a "maximum variation sample" (p. 17) would be helpful in interpreting the results. What was the justification for completing 30 interviews rather than any other number? It might be helpful to have Table 1 as an appendix, and to include details of ethnicity, and a general overview of the sample in the methods section.

We have provided further details regarding recruitment procedures and sample variation. We have opted not to move the demographics table to an appendix as experts in the field would expect to see such details in the main results section. The number of 30 participants, as with many qualitative studies, was based on a balance between a number that enabled a range of participants and experiences, an element of data saturation (although this is a debatable concept), enabling each participant’s unique story to be incorporated and resource allocation.

3) Data generation. Why were interviews undertaken rather than eg focus groups? Why were only patients interviewed rather than also including family carers and GPs or other health professionals? I would have liked more detail of and explanation for the use of pictorial representations (or a reference for their use), and more detail on how the individual interviews were conducted. Were negatice cases

sought?

There is a severe paucity of evidence on the experience of patients living with this rare disease – interviews were chosen to enable each participant to tell their own story in great depth.
This is the first in-depth study to explore the experience of patients living with PH – we certainly acknowledge the value of including health care professionals and this is currently being considered by the research team for a subsequent study. We were not expecting such vivid stories of the pre-diagnostic phase and the limitations and struggles faced by patients in relation to health services so the inclusion of HCPs was not an initial aim of the study. We have now alluded to this in the limitations section. Five interviews were conducted with a family member present but they provided no comments relevant to this paper – only encouraging body language and words such as, “yes that’s right”. Again, we acknowledge the value of including family members in the actual interview process in any subsequent study.

Not sure what extra detail the reviewer is requesting for conduct of the interviews – it is stated in the paper that these were conducted in the participants home, tape-recorded, lasted up to 90 minutes, and were semi-structured (sample questions provided). We have now made reference to the literature for the use of the patient journey data.

4) Data analysis. More detail is needed on how, when and by whom the data was analysed and synthesised. Were any software packages used? How did these themes develop (rather than others that you mention such as "frustration, anger and uncertainty" p.8)?

We have now added further details regarding data analysis. As sated on page 8, the words frustration, anger and uncertainty were present throughout the stories from early symptoms to early post diagnosis. Since Anderson’s model was used to help place the narratives into context main themes extracted represent the chronological journey as told by the participants.

5) Results and discussion. The results section is well laid out with a good use of illustrative quotations from a range of participants. The discussion section could then consider these accounts in comparison to those known from the literature for people with a range of other conditions, in order to highlight what is similar/different in a journey to diagnosis with PAH, and what the implicatipons of this may be, and what research is needed. Andersons' model could be used in this section.

Thank you for this comment but we feel that this has been addressed in the discussion. Reference is made to the trajectory of patients with COPD and cancer. In particular, reference is made to the experience of patients with ovarian cancer which also present with salient symptoms and how Anderson’s model has been applied to that population. The challenge with PAH is that very young children through to older people are affected by the condition – our age range is from 26 – 80 – this is a point that we have now discussed and hope that this is satisfactory.

6) Strengths and limitations. Given the claims made regarding the role of medical professionals in the patients' journey to a diagnosis of PAH, the exclusion of their views from the study has implications for this and future research. See also above on sampling strategy. The reservations expressed regarding the nature of qualitative data perhaps convey a misapprehension of what qualitative research can do. Working, as it can only do, with the pre-interpreted domain of understanding, its strength lies in its ability to explain people's perceptions and understandings of their experience, and the meanings they attach to it, which are what health professionals have to work with, regardless of "what really happened". Consequently I feel the method was appropriate to the research question, not a limitation on it, and contributed to its strength in starting to produce research knowledge of this area of patient experience.

We fully agree with this comment and thank you for providing such insight. We have made reference to the implications for further research as suggested.

7) Conclusions. In the abstract the first line should be replaced with a sentence answering the research question ie describing the patients experience of the trajectory, and pulling out the main implications of it for future research and consideration (since it can be problematic to make the leap from one qualitative study to recommendations for changes in policy and practice).

Thank you for this suggestion – we have revised the abstract to better reflect this.

VERSION 2 – REVIEW

REVIEWER	Marilyn Kendall Senior Research Fellow Centre for Population Health Sciences University of Edinburgh UK No competing interests
REVIEW RETURNED	06/03/2012

GENERAL COMMENTS	Thank you for the opportunity to review this revised manuscript. I am pleased to see that the authors have fully addressed the previous reviewers comments, especially with regard to describing and justifying the methods used and the place of this data in the overall study. I feel this has made the paper stronger, clearer to read and the results easier to interpret. On this b asis I would be happy for it to be published